Central Pleomorphic Adenoma of Mandible Mimicking Ameloblastoma – A Rare Case Report

Ravindra Manerikar[1], Amit Mani[2], Manas Bajpai[3], Shyam Reddy Karakalla[4]

[1] Department of Orthodontics and Dentofacial Orthopaedics, Rural Dental College, Pravara Institute of Medical Sciences, Loni (Maharashtra), India;
[2] Department of Periodontology, Rural Dental College, Pravara Institute of Medical Sciences, Loni (Maharashtra), India;
[3] Department of Oral Pathology and Microbiology, Rural Dental College, Pravara Institute of Medical Sciences, Loni (Maharashtra), India;
[4] Kamineni Hospital, Vijayawada (Andhra Pradesh), India

Received November 20, 2023; Accepted May 7, 2024.

Key words: Pleomorphic adenoma – Salivary gland neoplasm – Mandible – Jaw neoplasms

Abstract: Salivary gland neoplasms account for 3% of all head and neck tumours. Pleomorphic adenoma (PA) is the most common salivary gland tumour that mainly occurs in the parotid gland, followed by minor salivary glands of the oral cavity, however, the occurrence of PA inside the jaw bones is exceedingly rare and very few cases have been reported in the literature. Inside jaw bones these lesions tend to imitate large osteolytic lesions encompass a diagnostic challenge. An exhaustive review of the literature revealed only 10 cases of central pleomorphic adenoma. We present a rare case of primary PA that occurred inside the mandible and was provisionally diagnosed as ameloblastoma.

Mailing Address: Dr. Manas Bajpai, Department of Oral Pathology and Microbiology, Rural Dental College, Pravara Institute of Medical Sciences, Loni 413736 (Maharashtra), India; e-mail: dr.manasbajpai@gmail.com

https://doi.org/10.14712/23362936.2024.14

Introduction

Salivary gland tumours account for 3% of all head and neck tumours (Brookstone et al., 1992). Pleomorphic adenoma (PA) is the commonest salivary gland tumour, mainly occurring in the parotid gland and followed by minor salivary glands, although rare salivary gland tumours also occur in the jaw bones (Ojha et al., 2007).

Within the jaw bones mucoepidermoid carcinoma is found to be the most common tumour followed by adenoid cystic carcinoma and acinic cell carcinoma (Ojha et al., 2007; Bajpai et al., 2018; Manola et al., 2019). Occurrence of PA as primary central salivary gland tumour is rare and very few cases have been reported in the literature (Aghaghazvini and Aghaghazvini, 2015). We report a case of a large multilocular, osteodestructive lesion that was provisionally diagnosed as ameloblastoma and finally diagnosed as central PA after microscopic evaluation. We also describe the treatment modality performed in the present case.

Case report

An otherwise healthy 34-years-old man presented to a private dental clinic with the chief complaint of swelling and pain on the left, lower front region of his jaw for 8 months. The swelling was initially small and unnoticeable but gradually it started increasing and reached the current size. In the previous few days patient was suffering with dull pain in the same region. The personal history, family history,

Figure 1 – Extra-oral examination reveals asymmetry on the left side of face (black arrow denoting the swelling area).

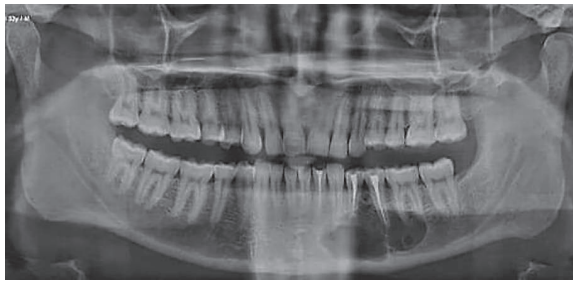

Figure 2 – Panoramic radiograph shows a large multilocular lesion of the left mandible.

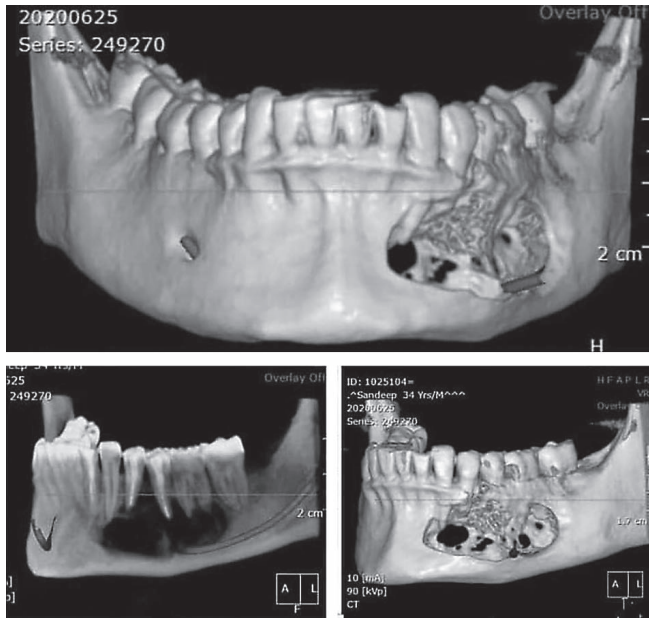

Figure 3 – 3D cone beam computed tomographic image exhibits perforations of buccal and lingual cortical plates.

and past medical history of the patient were found to be non-contributory to the presenting symptoms. Extra-oral examination revealed asymmetry on the left side of the face (Figure 1) on palpation the swelling was found to be hard and non-fluctuant, the overlying skin of the swelling had the same colour as that of the adjacent skin.

Intra-orally it revealed a solitary swelling extending from #33 to #36, on palpation it was found to be firm and tender. No paresthesia was noted in the initial examination. The colour of the mucosa was similar to the adjacent mucosa without any sign of ulceration. A panoramic radiograph revealed a large multilocular radiolucent lesion extending from #32 to #36, the roots of #34 and #35 and the mesial root of #36 were entrapped within the lesion (Figure 2). #32, #34 and #35 were root canal treated. A 3D reconstruction of the cone beam computed tomographic (CBCT) image revealed multiple buccal and lingual perforations (Figure 3).

After the correlation of clinical and radiographic features, a provisional diagnosis of ameloblastoma was given with the differential diagnosis of odontogenic keratocyst. An incisional biopsy was performed under local anesthesia and the specimen was sent for histopathological evaluation.

Hematoxylin and eosin-stained soft tissue sections revealed typical ductal and myoepithelial cells arranged in sheet patterns with multiple duct-like structures filled with eosinophilic coagulum (Figure 4). The nests of ductal and myoepithelial cells are interspersed with areas of hyalinization (Figure 5). Areas of extensive squamous metaplasia were also noted (Figure 6).

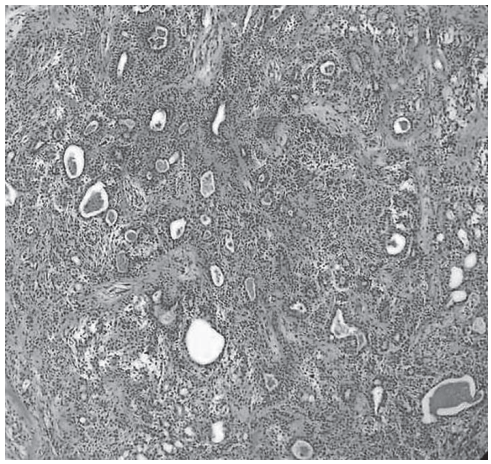

Figure 4 – Ductal and myoepithelial cells arranged in sheet patterns with multiple duct like structures filled with eosinophilic coagulum (hematoxilin and eosin stain 20×).

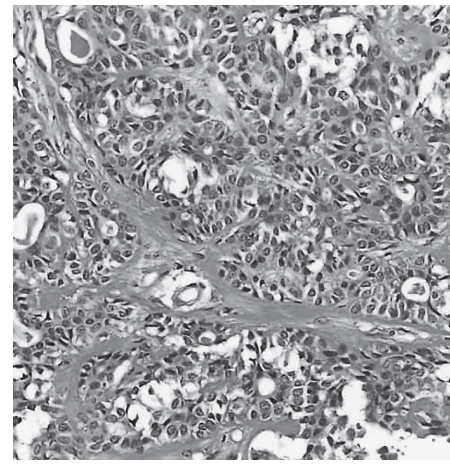

Figure 5 – Nests of ductal and myoepithelial cells are interspersed with areas of hyalinization (hematoxilin and eosin stain 40×).

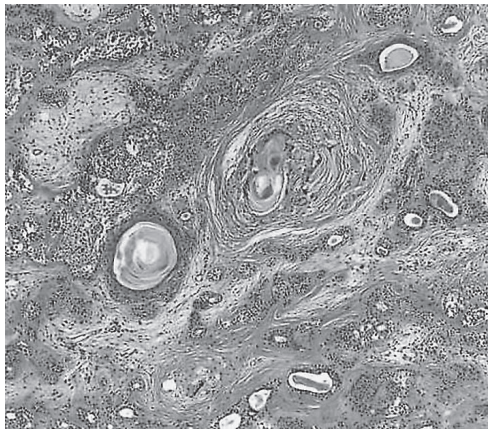

Figure 6 – Areas of extensive squamous metaplasia in the form of keratin pearls (hematoxilin and eosin stain 40×).

A final diagnosis of PA was given. A conservative treatment approach was opted for considering the age of the patient (young). Teeth #35 #36 and #37 were extracted and enucleation of the tumour was performed under general anesthesia, extra-oral approach was opted (Figure 7) because, with the extraction of three teeth and multiple perforations in the buccal and lingual plate, mandible would be susceptible to fracture due to the opposite pull suprahyoid and pterygomassetric sling, so it was decided to place the recon plate to strengthen the mandible, which would have been difficult to place intraorally (Figure 8). The patient was recalled every six months and the follow-up of the patient was uneventful and showed no recurrence to date (Figure 9).

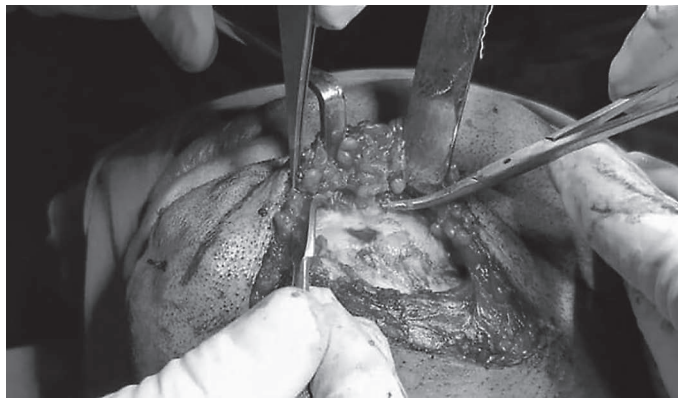

Figure 7 – Removal of the tumour via extra oral approach.

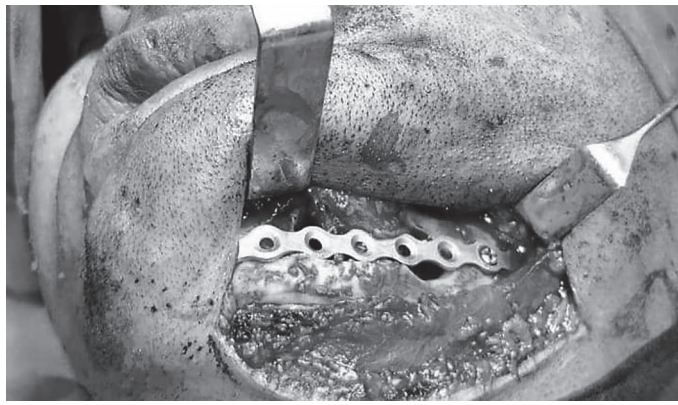

Figure 8 – Insertion of reconstructive plates.

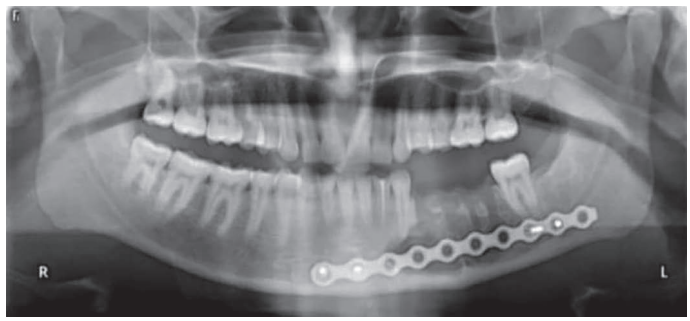

Figure 9 – Follow-up panoramic view reveals no sign of recurrence.

Discussion

The occurrence of salivary gland tumours as an intra-osseous/central tumour is a rare phenomenon. Mucoepidermoid carcinoma is the most common salivary gland tumour that has been reported to occur centrally, followed by adenoid cystic carcinoma (Alshagroud et al., 2017). PAs are the most common salivary gland

tumours that can occur in major and minor salivary glands. Intra-osseous PAs are extremely rare only 10 cases have been reported so to the best of our knowledge (Aver-De-Araujo et al., 2002). Several theories have been proposed to explain the intra-osseous origin of salivary gland tumours. The first theory explains the neoplastic changes in the ectopic glandular tissue inside the bone may give rise to intra-osseous salivary gland tumours, and the second theory explains that such tumours may arise due to the metaplastic changes in a pre-existing odontogenic cystic lining (Bouquot et al., 2000). The present case favours the first theory since it did not show any association with an odontogenic cyst. The mean age of occurrence for central PA is 58.8 (Arcuri et al., 2011). A rare case of central PA occurring in an eleven-year-old boy has also been reported (Bajpai, 2018). Radiologically central PA can predispose a diagnostic challenge to clinicians owing to their diverse radiographic features ranging from a unilocular cystic pattern to a multilocular destructive pattern (Arcuri et al., 2011). The present case showed a multilocular radiolucent pattern similar to ameloblastoma. Histopathologically, central PA shows features quite similar to the conventional PA of major and minor salivary glands (Aver-De-Araujo et al., 2002; Alshagroud et al., 2017). PA with extensive keratinization has also been reported in the literature (Bajpai and Pardhe, 2018), present case also showed squamous metaplasia at a few places. Wide local excision with the removal of the involved periosteum is considered as the treatment of choice though the present case was treated by the thorough enucleation of the mass (Vicente et al., 2008). Regarding why the present case was treated with enucleation rather than resection because we felt that the patient was young and could not be left without a reconstruction, where free fibula was an obvious choice, but that itself could have its possible complications, the patient chose to go conservative with organ preservation, moreover, the peripheral osteotomy was performed to reduce the chance of recurrence.

Recurrence rate of 2–44% has been reported in the parotid gland PA (Kowalik, 1966). There is a paucity of information regarding the recurrence rate of intra-osseous PA.

Conclusion

Central Pleomorphic adenoma is the rarest diagnostic possibility that comes to the mind of a clinician when comes a patient with an osteodestructive central lesion. Such lesions can impose a diagnostic difficulty due to their rarity in jaws and overlapping clinical and radiological features. Centrally located Pleomorphic adenoma should be followed up for a long time because their recurrence rate and malignant transformation have not been sufficiently described yet.

References

Aghaghazvini, L., Aghaghazvini, S. (2015) Intraosseous pleomorphic adenoma of the mandible. *Iran. J. Radiol.* **12(1)**, e6921.

Alshagroud, R., Kamoh, A., Popat, S. R., Brandwein-Weber, M., Aguirre, A. (2017) Metastasizing pleomorphic adenoma case report and review of the literature. *Head Neck Pathol.* **11(4)**, 487–493.

Arcuri, F., Grivetto, F., Poglio, G., Brucoli, M., Benech, A. (2011) Simultaneous palatal pleomorphic adenoma and mandibular keratocystic odontogenic tumor in a pediatric patient. *J. Craniofac. Surg.* **22(6)**, 2347–2350.

Aver-De-Araujo, L. M., Chaves-Tarquinio, S. B., Neuzling-Gomes, A. P., Etges, A. (2002) Intraosseous pleomorphic adenoma: Case report and review of the literature. *Med. Oral* **7(3)**, 164–170. (in English, Spanish)

Bajpai, M. (2018) Central pleomorphic adenoma of mandible – Report of the first case in a pediatric patient. *National Journal of Health Sciences* **3**, 63–65.

Bajpai, M., Pardhe, N. (2018) Pleomorphic adenoma of submandibular gland with extensive cystic keratinization. *J. Coll. Physicians Surg. Pak.* **28(1)**, 84.

Bajpai, M., Pardhe, N., Chandolia, B., Arora, M. (2018) Central acinic cell carcinoma of the mandible simulating as benign odontogenic lesion: A case report. *Iran. J. Med. Sci.* **43(2)**, 223–226.

Bouquot, J. E., Gnepp, D. R., Dardick, I., Hietanem, J. (2000) Intraosseous salivary tissue: Jawbone examples of choristomas, hamartomas, embryonic rests and inflammatory entrapment. *Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.* **90**, 205–217.

Brookstone, M. S., Huvos, A. G. (1992) Central salivary gland tumors of the maxilla and mandible: A clinicopathologic study of 11 cases with an analysis of the literature. *J. Oral Maxillofac. Surg.* **50(3)**, 229–236.

Kowalik, S. (1966) Mixed tumor starting from the jaw bone. *Otolaryngol. Pol.* **20**, 563–564. (in Polish)

Manola, M., Moscillo, L., De Luca, E., Lalinga, A. V., Possidente, L. (2019) Intraosseous pleomorphic adenoma of the maxilla: A case report and literature review. *J. Otolaryngol. Rhinol.* **5**, 064.

Ojha, J., Bhattacharyya, I., Islam, M. N., Manhart, S., Cohen, D. M. (2007) Intraosseous pleomorphic adenoma of the mandible: Report of a case and review of the literature. *Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.* **104(2)**, e21–e26.

Vicente, O. P., Marques, N. A., Aytes, L. B., Escoda, C. G. (2008) Minor salivary gland tumors: A clinicopathological study of 18 cases. *Med. Oral Patol. Oral Cir. Bucal* **13**, 582–588.